

A New Concept in the Treatment of Oral Venous Malformation using Sclerotherapy

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ABSTRACT

Introduction: Vascular malformations accounts for approximately 7% of all benign tumors, the majority are seen in the head and neck region. Vascular malformations (VM) are a general term used to describe a group of lesions formed by an anomaly of the capillary, venous, lymphatic, and arterial system.

Case report: We present a case of localized dome-shaped swelling measuring approximately 4 cm × 3.5 cm in the right buccal mucosa opposite to cusp of 1st premolar and 2nd molar. Lesion around 2-3 mm below parotid duct opening, bright red in color with irregular pebbled surface, and the surrounding area appeared pale pink in color, no visible pulsation. On palpation, it was soft to firm in consistency, immobile, and nontender. There was no apparent discharge nor palpable thrill and bruits, but the swelling blanched on compression with a glass slide. Based on the history, clinical features and color Doppler ultrasound (US), it was diagnosed as venous malformation.

Conclusion: The management of vascular lesions is challenging. Though there are various treatment options available but sclerotherapy, proves to be an effective way to treat them with limited amount of armamentarium, cost effectiveness and minimal complication.

Keywords: Oral Venous Malformation, Sclerotherapy

CASE REPORT

A 55-year-old male patient presented in the department of dental surgery at MGM government medical college and hospital Jamshedpur, with a swelling involving the right half of the buccal mucosa which was there since childhood, gradually increased in size for the past 20 years. Patient did not give any history of pain, neither any difficulty in speech and swallowing. Past medical, dental, and family history was noncontributory. On intraoral examination, a localized dome-shaped swelling measuring approximately 4 cm × 3.5 cm was present in the right buccal mucosa opposite to cusp of 1st premolar and 2nd molar. Borders of the swelling were well defined, and it extended from the distal cusp tip of first premolar to distal of second molar. Lesion was around 2-3 mm below parotid duct opening. It was bright red in color with irregular pebbled surface, and the surrounding area appeared pale pink in color [Figures 1-3]. Pulsation was not felt and on palpation, it was soft to firm in consistency, immobile, and nontender. There was no apparent discharge nor palpable thrill and bruits, but the swelling blanched on compression with a glass slide.

Based on the clinical findings, a working diagnosis of arteriovenous malformation (AVM), hemangioma, lymphangioma and hemorrhagic cyst was arrived following which necessary radiologic investigations were carried out. Color Doppler ultrasound (USG) revealed an ill-defined heterogenous lesion present in the right anterior, lateral, and posterior aspect of the mid half of the buccal mucosa [Figure 4 and 5]. The lesion was highly vascular with no feeder vessel, flow was predominantly venous.

Considering the provisional diagnosis, unwillingness of the patient for surgery, management with intralesional injection of 30 mg/ml of sodium tetradecyl sulfate (STS) was decided with the written consent of the patient.

Technique of injection

Surface anesthesia was given by 15% xylocaine spray. 1 ml of 3% sodium tetradecyl sulphate was injected intralesionally at multiple sites into the mucosa, first at the periphery and then

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into the centre of the lesion with insulin syringe, contained within the lesion using manual compression for 5 to 10 minutes to ensure stasis wherever it was possible [Figures 6 and 7]. Total dosage was not exceeded by more than 2.0 ml and care was taken to prevent extravasation of sclerosant. Post sclerotherapy, antibiotics, analgesics and anti-trypsin preparations were given along with ice-pack applications. The injections were repeated after an interval of two weeks, total three sessions of injections were given at an interval of two weeks.

Sclerotherapy provided significant improvement and resolution of the lesion. Six weeks of follow up period was there. During second week of follow up period, mucosal ulceration and sloughing was seen with mild burning sensation, which subsided gradually. On fourth week follow up, lesion had shrunken to huge extent, ulcerated margin had healed, mild fibrosis was palpated at the injection site. On fifth week follow up, lesion had completely resolved and normal pink mucosal color was seen with minimal fibrosis at the site [Figures 8-10].

Complications and sequelae

The patient experienced pain and swelling to a variable degree which lasted typically for a week. A mild degree of inflammation of the overlying mucosa was seen with mucosal ulceration, sloughing and burning sensation, which healed with local wound care.



Figure-1: right profile of the patient



Figures-2 and 3: intraoral appearance of the lesion



Figures-4 and 5: Colored ultrasonography shows ill-defined heterogeneous lesion, highly vascular with no feeder vessels, predominantly venous

DISCUSSION

International Society for the Study of Vascular Anomalies Classification divides vascular anomalies into two primary biological categories: (1) Vasoproliferative, vascular neoplasms and (2) Venous malformation. Vascular malformations are true in-born errors in the embryologic development of the vascular tree, and by definition are all present at birth though not all clinically apparent. VM occur approximately in 1% of births but majority of the patients do not turn out for the treatment.² These lesions occur at any area of the body; gradual in onset and progression, most common sites in the oral cavity are anterior two-thirds of the tongue, palate, and gingiva and buccal mucosa. In this case, lesion was seen in the buccal mucosa since childhood, and it was gradually increasing in size which can be correlated with the features of arterio-venous malformation. Vascular malformations in the oral cavity leads to discomfort and potentially serious clinical problems. Lesions involving buccal mucosa, tongue can cause problems such as recurrent hemorrhage, biting of the lesion, pain, and difficulty with



Figure-6: sodium tetradecyl sulphate 3% and an insulin syringe



Figure-7: Administering the solution first at the periphery and then into the center of the lesion; **Figure-8:** two week follow up: reveal decrease in bulk of the lesion, mild ulcerated margin seen



Figure-9: Fourth week follow up: shows healing ulcer and moderate reduction in size of the lesion; **Figure-10:** fifth week follow up: complete disappearance of the lesion, mild fibrosis over the injection site was palpable

speaking, mastication or deglutition.³

Various treatment options for small and peripheral vascular lesions are conventional surgical excision, laser therapy, cryotherapy, selective embolization, sclerotherapy, and medical treatment using beta blocker or steroid.⁶

The lesions which are larger and deeper are treated using embolization in which obliteration of adjacent vessel is required. These treatments leads to irreversible tissue injury and gradual fibrosis, currently, surgical resection is considered as the best treatment modality for vascular benign lesions which occur in head and neck region. However, surgical resection is limited when complete resection is not possible, where it may cause critical bleeding or where crucial organs can be injured. Extensive surgery in the oral cavity can induce difficulty in mastication.

Lesions that are small or located at the site where esthetic is a concern, sclerotherapy can be an alternative to surgical treatment.⁷ Sclerotherapy can regress lesions partially or entirely and is effective for relieving symptoms. Furthermore, the procedure is simple, less invasive, and inexpensive. Using sclerosant, it is possible to receive treatment as an outpatient basis.^{3,4} However, sclerotherapy should be performed with care, because it can cause complications such as pulmonary embolism, anaphylaxis, nerve damage, increased pain, and disseminated intravascular coagulation, tissue necrosis, and sloughing (4%) and airway compromise (1%).⁷

Various Sclerosing Agents- Ethanol preparations suitable for sclerotherapy, include 95% to 98% of dehydrated forms, generally available through hospital pharmacies used for neurolysis. It rapidly denatures proteins in the endothelial lining of vessels and, in stagnant channels, which results in immediate thrombosis. Ethanol also causes neurolysis locally. Systemic effects include CNS depression, hypoglycemia, hyperthermia, cardiac arrhythmias, pulmonary vasoconstriction and pulmonary hypertension, and electromechanical dissociation. To avoid this, ethanol should be administered in small dosage, with adequate time for recovery between injections. Maximum dosage of 1 ml/kg should not be exceeded in one procedure.

Detergent Sclerosants- This category includes sodium tetradecyl sulfate, the most commonly used sclerosant followed by ethanolamine and polydocanol. Like alcohol, these sclerosants damage the endothelial lining and lead to coagulation of the intraluminal blood products. Detergent sclerosants can be opacified with water-soluble contrast medium or with oily contrast medium, addition of air results in a microfoam form which is more effective than the bland solution. A reasonable dose limit for image-guided sclerotherapy is 0.5 ml/kg.

Sodium tetradecyl sulfate has been used as a sclerosant from many decades with a high level of safety and efficacy. Chemically, this drug is unique into itself and unrelated to other commonly used sulfur-containing pharmacologics that normally have a strong allergic potential.⁸ It works by creating a localized inflammatory reaction which leads to obliterative thrombosis of hemangiomatous space that result in subsequent fibrosis of the endothelial spaces, and finally regression of the lesion without affecting the bone.⁹

Literature suggest few cases of anaphylactic reaction and pulmonary embolism that were seen following intravenous

injection of STS.⁸ Minkow *et al.*, in their study using intralesional injection of 3% (STS) in intraoral hemangioma showed satisfactory results in all patients with minimum side effects whereas in another study on venous malformations using 3% STS, complications occurred¹⁰ similar to our case.

Other Sclerosants - Doxycycline which is available as a powder can be suspended in saline or contrast medium. It is often used for sclerotherapy of lymphatic malformations. It is painful to inject, but effective and relatively nontoxic.

Bleomycin has been used for decades in the sclerotherapy of lymphatic malformations (LM). It has some systemic side effects which include pulmonary fibrosis, hair loss and pigmentation, therefore the quantity of this drug used in each session must be carefully limited.¹⁴ Large quantities of sclerosants can result in hemoglobinuria which require aggressive hydration and urine alkalinisation.

Different preparations of sclerosing agent are used these days one of them are sclerosant foam. These were first developed by Tessarri in 2001¹¹, use of which has shown considerably faster rate of healing and less amount of post-operative complication.¹⁵ Neodymium yttrium-aluminium-garnet (Nd:YAG) laser therapy has also been increasingly used when venous lesions are small, located in difficult anatomical situations, and have not responded to other treatments with good control of VMs.¹⁷ It is usually administered via a flexible optic cable and causes the photocoagulation of blood vessel tissue. Its effects are highly localized which can cause adverse effects, especially if important structures, such as nerves are too close to the area of action.^{6,17} When they are used with appropriate precautions, it is a fast, safe, and easily administered modality.

CONCLUSION

The management of vascular lesions is challenging. Though there are various treatment options available but sclerotherapy, proves to be an effective way to treat them with limited amount of armamentarium, cost effectiveness and minimal complication.

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